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Preauricular Sinus-classic versus Variant Types among a Cross-section Subjects in Southwestern Nigeria

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Authors' contributions

This work was carried out in collaboration between both authors. The author SKA designed, analyzed, interpreted and prepared the manuscript. Author OW arranged the references. Both authors read and approved the final manuscript.

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ABSTRACT

Background: The variant types of preauricular sinuses ('postauricular sinuses') presenting as postauricular swelling have rarely been reported. Preauricular sinuses and cysts result from developmental defects of the first and second pharyngeal arches. This and other congenital ear malformations are sometimes associated with renal anomalies such as branchio-oto-renal syndrome. The variant types of preauricular sinuses ('postauricular sinuses') presenting as postauricular swelling have rarely been reported.

Aim: The study is a prospective one carried out to find out the prevalence of preauricular sinus (classic and variant) in our community.

Methodology: This was a prospective study carried out between June 2013 and May 2018 on patients seen in Ear, Nose and Throat department of the Federal Teaching Hospital, Ido-Ekiti, as well as a cross-section of population in different communities of Ekiti state, Nigeria, after obtaining ethical approval from the hospital ethical committee and informed consent was taken from all subjects enrolled in the study. Detailed clinical history, administered interviewer's assisted questionnaires full examination and. Data obtained were collated and analyzed.

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Results: Over the five (5) year study period, a total of 4,350 subjects were seen both within the hospital and the cross sections of the Ekiti community. Out of these subjects, 108 were found to have preauricular sinus either classic or its variant. The prevalence is 2.48% (both Classic and Variants). Male accounted for 78(72.2%) of the total subjects while the female was 30(27.8%). The age of the subject range from 1 to 70 years with the mean age of 36.17 years. The preponderance was found in the age group 11-20 years (Table 1). Majority of the subjects were pupil/student (preschool and infants included) and accounted for 37.0%. Surgery was done for 25 consented subjects. 80.0% belong to the classic type and 20.0% variant type. Recurrence after surgery is noted to be higher in variant type (60.0%) as compared to the classic type.

Conclusion: Preauricular sinus, classic, and the variant is a congenital anomaly that is common in this part of the world our environment with a small proportion showing clinical symptoms. Prevalence was 2.48%, which is lower compared to some other African studies higher in variant type as compared to the classic type. Hearing loss was found in two of our studied subjects. Recurrence after surgery was noted to be higher in variant type as compared to the classic type.

Keywords: *Preauricular sinus; postauricular swelling; hearing loss; preauricular abscess.*

1. INTRODUCTION

A preauricular sinus (also known as a congenital auricular fistula, a congenital preauricular fistula, a Geswein hole, an ear pit [1], or a preauricular cyst [2]) is a common congenital malformation characterized by a nodule, dent or dimple located anywhere adjacent to the external ear [3]. The frequency of preauricular sinus differs depending on the population: 0.1–0.9% in the US, 0.9% in the UK, and 4–10% in Asia and parts of Africa [4].

They are congenital malformations that usually occur at the anterior margin of the ascending limbs of the helix of the external ear [5,6,7].

Preauricular sinuses are inherited features, and most often appear unilaterally. They are present bilaterally in 25–50% of cases. It is more often unilateral while bilateral forms occur usually as an inherited disorder and its association with deafness and as part of syndromic disorders had been reported [5,6,7].

Preauricular sinuses and cysts result from developmental defects of the first and second pharyngeal arches [8]. This and other congenital ear malformations are sometimes associated with renal anomalies [9]. In rare circumstances these pits may be seen in genetic conditions such as branchio-oto-renal syndrome; however, these conditions are always concurrent with other health concerns.

The variant types of preauricular sinuses ('postauricular sinuses') presenting as postauricular swelling have rarely been reported. Chang reported only three cases presenting as infected postauricular cysts in 2005 [10].

Postauricular masses or cysts located posterior to the External auditory canal are usually considered as inflamed lymph nodes, infected epidermoid cysts, sebaceous cysts, or branchial cleft cysts. These masses require antibiotics, incision, and drainage, or excision. However, there are some recurrent masses or swellings in the postauricular area, even if they are properly treated. One of the reasons for this is the variant type of preauricular sinuses. The variant type of preauricular sinuses is a 'postauricular sinus' whose sac is strangely located in the postauricular area and also presents the opening pit near the ascending limb of the helix [11].

The purpose of this study was to analyze the incidence/prevalence, histories, clinical manifestations, in a cross-sectional population of southwestern Nigeria.

2. MATERIALS AND METHODS

The study is a prospective one carried out on patients seen in Ear, Nose and Throat department of the Federal Teaching Hospital, Ido-Ekiti, as well as a cross-section of the population in different communities of Ekiti state, Nigeria.

The study was carried out between June 2013 and May 2018 after obtaining ethical approval from the hospital ethical committee and informed consent was taken from all subjects enrolled in the study.

All patients with clinical diagnosis of preauricular sinus and its variants were enrolled in the study, interviewer's assisted questionnaires were administered to all the subjects and detail

histories were taken. Thorough otologic, nasal and throat examinations were performed on all the subjects. Detailed general examinations were carried out to rule out associated congenital anomalies.

Audiologic assessment including pure tone audiometry, tympanometry, and an otoacoustic study was performed to assess the quality and quantity of hearing acuity. In addition, abdominopelvic ultrasound scan was performed to rule out congenital renal anomalies in patients identified with preauricular sinus.

The patients with preauricular sinus were categorized into two groups according to the position of the sinus sac; i.e., the classical group or the variant group. The classical group was defined as the group of patients with pits located on the (superior) anterior to the EAC, and the variant group as those whose sacs were located on the posterior side of the EAC.

The majority (N=15, 60%) of the patient had supraauricular approach while the minority (N=5, 40%) had the elliptical incision around the opening, after sinus tract identification with methylene blue or a probe, and followed by dissection of the tract.

The limitation of this study was there were few numbers of subjects with the condition but refused to give consent for the study and thus excluded.

Data obtained were collated and statistically analyzed descriptively using SPSS version 16 computer software. Data were expressed by using tables, bar and pie charts.

3. RESULTS

Over the five (5) year study period, a total of 4,350 subjects were seen both within the hospital and the cross sections of the Ekiti community. Out of these subjects, 108 were found to have preauricular sinus either classic or its variant. The prevalence is 2.48% (both Classic and Variants). Male accounted for 78(72.2%) of the total subjects while the female was 30(27.8%). The age of the subject range from 1 to 70 years

[Table 1] with the mean age of 36.17 years. The preponderance was found in the age group 11-20 years [Table 1]. Majority of the subjects were pupil/student (preschool and infants included) and accounted for 37.0% [Table 2]. Very few numbers, 2(1.9%) were retiree, 2(1.9%). Ninety-nine subjects (91.7%) of the subjects had the classic type of the preauricular sinus [Table 3] while 9(8.3%) had the variant type.

Table 1. Age distribution

Age range	Number (N)	Percentage (%)
1-10	25	23.1
11-20	40	37.0
21-30	18	16.7
31-40	7	6.5
41-50	9	8.3
51-60	6	5.6
61-70	3	2.8

Table 2. Occupation distribution

Occupation	Number (N)	Percentage (%)
Pupil/Student	40	37.0
Apprentice	23	21.3
Health Worker	3	2.8
Farmer	5	4.6
Business	10	9.3
Civil Servant	25	23.1
Retiree	2	1.9

[Table 3] relates the sex predilections with the classic and variant types and it showed that male sex (72 in classic, 6 in variant) was more affected than female sex (27 in classic, 3 in variant).

The same relationship is also illustrated in [Fig. 1]. A significant proportion (N=83,76.9%) were not symptomatic while 23.1% (N=25) showed symptoms [Fig. 2]. Majority of the symptomatic subjects were female (N=18,72%) and the right side was commonly implicated (N=15,60%) with the classic type of preauricular sinus having the highest proportion (N=22,88%) [Table 4]. Laterality was observed with right ear predominance (N=3835.2%) over left (N=30, 27.8%) [Fig. 3].

Table 3. Correlating the sex distribution to the classic and variant types of preauricular sinus

	Male	Female	Number (N)	Percentage (%)
Classic	72	27	99	91.7
Variant	6	3	9	8.3
Total: N(%)	78(72.3)	30(27.8)	108	100

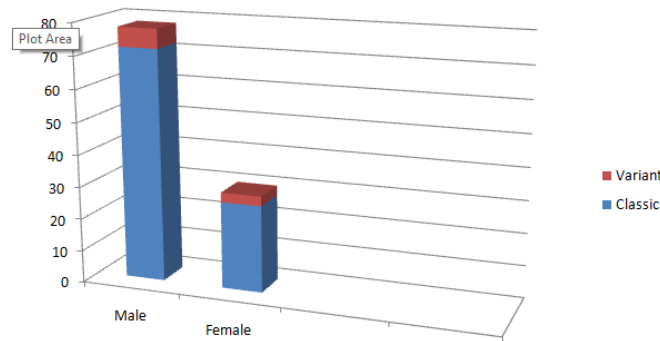


Fig. 1. Sex distribution of classic and variant pre-auricular sinus

Table 4. Correlating the symptomatic subjects with the sex distribution, symptomatic side and the type of preauricular sinus

	Sex (N,%)		Side (N,%)		Type (N,%)	
	Male	Female	Right	Left	Classic	Variant
Symptomatic	N=7	N=18	N=15	N=10	N=22	N=3
Subjects(N=25,%=21.3)	%=28	%=72	%=60	%=40	%=88	%=12

Table 5. Symptom Distribution among the subjects (More than one symptoms are present in some subjects)

Symptoms	Number (N)	Percentage (%)
Recurrent Ear Pain	83	41.3
Recurrent Ear Discharge	38	18.9
Preauricular/Post auricular Abscess	25	12.4
Preauricular/Postsuricular Swelling	38	18.9
Preuricular/Postauricular Erythema	15	7.5
Hearing Loss	2	1.0
Total	201	100

Table 6. Recurrence rate

Preauricular sinus	Number(N)	Recurrent (N,%)	Non-recurrent (N,%)
Classic	20	5(25)	15(75)
Variant	5	3(60)	2(40)
Total	25	8(32)	17(68)

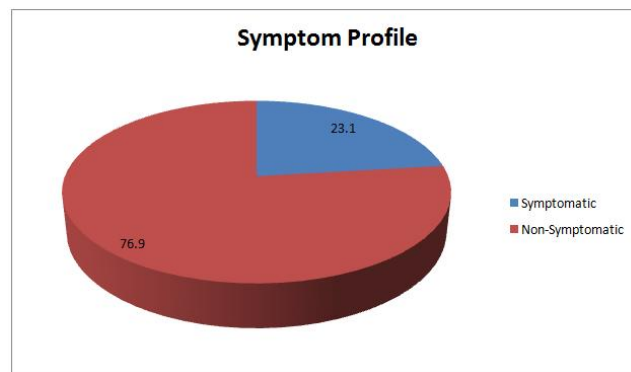


Fig. 2. Percentage illustration of symptomatic to non-symptomatic subjects

Commonly presented symptoms were recurrent ear pain (N=83,41.3%), recurrent ear discharge (N=38,18.9%) and preauricular abscess (N=25,12.4%). Preauricular erythema (N=15, 7.5%) and hearing loss (N=2,1.0%) made a small proportion [Table 5].

Surgery was done for 25 consented subjects, 20(80.0%) belong to the classic type and 5(20.0%) variant type [Table 6]. Recurrence after surgery is noted to be higher in variant type (60.0% of N=5) as compared to the classic type (25.0% of N=20), though classic had a higher number(N=20) of operated subjects [Table 6]. Also, it was noted that the recurrence rate was higher in patients who have an elliptical incision approach (N=6,75%) as compared to the supraclavicular approach (N=4,25%).

4. DISCUSSION

Preauricular sinuses were first described by Heusinger in 1864. They are a common congenital anomaly in children [12]. Most preauricular sinuses appear externally as a small opening near the anterior margin of the ascending limb of the helix, and they are classically located at the area anterior to the external auditory canal (EAC). They are frequently uncovered on routine physical examination; 0.1% to 0.9% in the United States, [13] 4% to 6% in Asian, [14,15] and 4% to 10% in some areas of Africa [16]. Preauricular sinuses originate from a defective or incomplete fusion of the six auricular hillocks [17].

The preauricular sinus is a very common congenital anomaly with an incidence range from 0.1% to 10%, depending on the area or people. It has been generally known that preauricular sinuses are more common among African or Asian than in white people [12,13,14,15]. Our study showed a prevalence of 2.48% (both Classic and Variants). Many European and American reports showed a similar prevalence of preauricular sinus which was much lower than findings from African countries [5,7,18]. This is because most of the African studies did not include the variant type which is relatively rare.

Most preauricular sinuses are located in the superior-anterior to the EAC and have opening pits just anterior to the ascending limb of the helix. This is the classic type. The variant types of preauricular sinuses ('postauricular sinuses') presenting as postauricular swelling have rarely been reported, we, however, identified one case

of postauricular sinus presenting as postauricular swelling in this study, [Fig. 5]. Chang reported only three cases presenting as infected postauricular cysts in 2005 [10] and Yeo reported 8 cases among 206 ears with the preauricular sinus that had pits located in the cyma concha and two cases located in the postauricular area [19]. However, the variant types of preauricular sinuses, 'postauricular sinuses' seem to be more common. Seong et al. [11] diagnosed 11 cases (10.9%) of the variant types out of the 101 patients with preauricular sinuses. Ninety-nine subjects (91.7%) of the subjects in our study had the classic type of the preauricular sinus [Table 3] while 9(8.3%) had the variant type, similar to the findings by Seong et al. [11]. Therefore, many cases of the variant types of preauricular sinuses among the postauricular swellings or abscesses appear to have been missed or ignored in many studies. The higher incidence of previous operation histories in the variant group compared with the classical group is evidence of this possibility.

The variant types of preauricular sinuses could be classified into three types according to the location of pits [Fig. 4]: pits located on the middle area on the crus (type 1), pits located on the superior area of the crus (type 2), and pits located on the cyma concha (type 3) [11]. Two subjects with type 3 were seen in our study. Physicians need information about the variant type of preauricular sinuses that presents with postauricular swelling as well as the variants of it to avoid misdiagnosis and mismanagement.

Preauricular sinuses are generally known to affect men and women equally, and the results shown in Seong et al's study are similar (44 male, 46 female) to this, although some studies have shown more frequent occurrence in women [20,21,22,23]. On the other hand, the variant types of preauricular sinuses were more common in male patients than female patients (7 males, 4 females) [11]. However, our study showed a higher proportion of male (N=78,72.2%) than female (N=30,27.8%) which is similar to the findings by Adegbiyi et al. [18]. Jimoh et al's [24] study revealed a male preponderance. Affection of the right ear is commoner in our study than the left. This may be because many people are of right-handedness than left-handedness. So the right ear is more probed than the left ear. There are studies which showed that men and women were equally affected [25,26,27]. Some works support women to be predominantly affected [28,29,30].

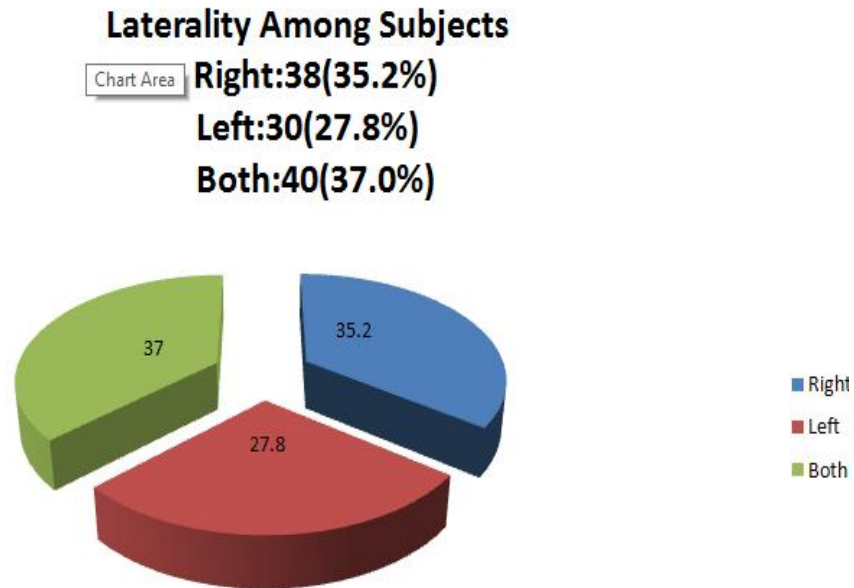


Fig. 3. Laterality of preauricular sinus among the subjects

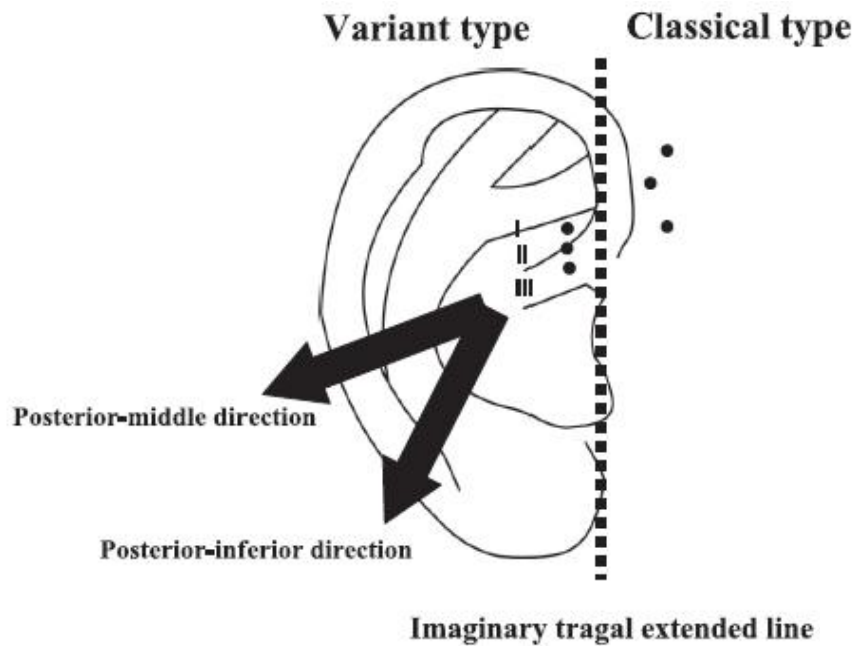


Fig. 4. (Choi et al. 2007): Illustrates the fistular pits of the variant type of the preauricular sinuses are located on the posteriorly to imaginary tragal extended line (dotted line) connecting between the tragus and the posterior margin of the ascending limb of helix. The variant types are classified into three types according to the location of pits: pits located on the middle area on the crus (type 1); pits located on the superior area of the crus (type 2); and pits on the cymba concha (type 3)



Fig. 5. Postauricular sinus presenting as postauricular swelling

Other studies also establish commoner unilateral and higher right ear cases [30]. The preauricular sinuses were identified in both ears of about 20% of the classical and variant types, respectively, [11] supporting what our study established [Table 4].

The variant types of preauricular sinuses could be classified into three types according to the locations of pits [Fig. 4]. Type 1 (pits located on the middle area of the crus) was most common and easily detectable; however, types 2 and 3 were very difficult to identify because the pits were sheltered in the helix and antihelix. The direction of the fistular pits of the variant types was usually posterior middle to the postauricular area, and there were no tracts parallel to the EAC or openings in the canals, suggesting that these lesions are a kind of preauricular sinuses with the postauricular presentation, rather than type I cyst of the first branchial cleft.

Various theories regarding its development had been documented [30,18,31]. Notable among these theories include; an incomplete fusion of auricular hillocks during the embryonal period or incomplete closure of the dorsal part of the first pharyngeal groove [31,32,33]. Some physicians believed that preauricular sinus is important markers of teratogenic exposure [34,35].

In Africans, preauricular sinus is associated with strong cultural belief that militates against its prompt treatment [34].

Most people with this malformation are asymptomatic as evident by our study [Fig. 2]

and many African and European studies. Once infected, these sinuses are rarely asymptomatic, and often develop recurrent acute exacerbations. Common clinical presentations of this condition, as evident by this study [Table 5], are recurrent ear pain (41.3%), discharge (18.9%), erythema (7.5%), preauricular swelling (18.9%) and hearing loss (1.0%). One or more symptoms are present in a single subject. Hearing impairment was found to be 8/1000 among infants with preauricular skin tags or ear pits in a study done by Daphne et al. [36].

Some people, however, may present with facial cellulitis or ulcerations located on the anterior side, contiguous to the ear [19]. Some patients present with chronic intermittent drainage of purulent material from the opening. Draining sinuses are prone to infection.

Although there is no necessity to treat patients with no clinical signs or symptoms, improper management of patients with infected sinus will result in recurrent infection and possibly severe postoperative scarring [37].

In the acute phase of the preauricular sinus infection, clinical management hinges on appropriate antibiotic therapy directed against the causative pathogen. Abscesses should be drained. Recurrent or persistent infection of preauricular sinus requires surgical excision of the sinus and its tract during the quiescent portion of the recurrent inflammatory cycle [5].

Surgery was done for 25 consented subjects, 80.0% of which belong to the classic type and

20.0% variant type [Table 6]. Recurrence after surgery was noted to be higher in variant type(60.0%) as compared to the classic type(25.0%), though classic had a higher number of operated subjects [Table 6]. These are uncommon reports as compared to many studies done before. The majority (N=15, 60%) of the patient in our current study had supraauricular approach while the minority (N=5,40%) had the elliptical incision around the opening, after sinus tract identification with methylene blue or a probe, and followed by dissection of the tract. It was also noted in our study that the recurrence rate was higher in patients who have an elliptical incision approach (N=6, 75%) as compared to the supraclavicular approach (N=4,25%). These results are in agreement with Prasad et al. [38] (5% recurrence) and Lam et al. [39] (3.7% recurrence rate) on using the supra-auricular approach for excision of preauricular sinus and also agree with the systematic review done by El-Anwar and El-Aassar [40] who found that the recurrence rate was 4/333 (1.2%) with the supra-auricular approach after reviewing nine pieces of research using this technique.

Few cases of hypertrophic scar, keloid and perichondritis were noted in this study.

Some other studies also revealed syndromes association with preauricular sinus [41,42]. Such associated conditions and syndromes are Branchio-oto-renal (BOR) syndrome, Treacher Collins syndrome, Hemifacial microsomia syndrome, Branchio-oto-ureteral syndrome, Branchi-otic syndrome, Branchi-otic-costal syndrome, Tetralogy of Fallot and Clinodactyly. Hearing loss although very rare was found in two of our studied subjects [41,24].

None of our studied subjects had any of these syndromes [5,7,43].

Some clinical conditions may appear like postauricular sinus/swelling and mislead diagnosing it. Such conditions include acute mastoiditis and postauricular lymphadenopathy.

5. CONCLUSION

Preauricular sinus, classic and variant, is a congenital anomaly that is common in this part of the world our environment with a small proportion showing clinical symptoms. Prevalence was 2.48%, which is lower compared to some other African studies. Hearing loss although very rare

was found in two of our studied subjects. Recurrence after surgery was noted to be higher in variant type as compared to the classic type.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard or university standard, written approval of Ethics committee has been collected and preserved by the authors.


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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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